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CLINICAL PROCEEDINGS

of the
CHILDREN'S HOSPITAL

WASHINGTON, D. C.



February 1953

VOLUME IX

NUMBER 2



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CLINICAL PROCEEDINGS

OF THE CHILDRENS HOSPITAL

13th and W Streets, Washington 9, D. C.

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DANGERS INCIDENT TO THE USE OF POLYETHYLENE TUBING IN FEEDING INFANTS

EDITORIAL

E. Clarence Rice, M.D.

Recent articles have set forth the advantages of using polyethylene tubing in feeding the premature infant. Since each of two articles^(1, 2) mention a death caused by this tubing perforating the stomach of intubated infants, and since members of the Department of Pathology at The Children's Hospital have performed necropsies on two babies in whom the insertion of the tubing either caused the death of the patient or contributed to it, it was deemed advisable to stress the dangers incident to the use of polyethylene tubing in feeding infants.

The pertinent findings in the two patients who were autopsied were: (1) perforation of the esophagus followed by acute mediastinitis and hemothorax, and (2) ulceration and necrosis of the esophagus found three days after the polyethylene tubing had been introduced into the esophagus and stomach. The latter patient had agranulocytosis and marked thrombocytopenia as a part of meningo-encephalitis and hepatitis, being a poor subject for a feeding tube to be allowed to remain in the esophagus for an extended period.

Kunz⁽²⁾ recommends the use of paraffin-tipped tubing to prevent perforating the stomach. Polyethylene tubing with bulbous tip is now available commercially or can be prepared by the physician by adding a paraffin tip. As an additional safeguard it is recommended that such tubing not be allowed to remain in the esophagus for several days' time because of the possibility of causing ulceration of the mucous membrane due to the presence of a foreign body. If these precautions are taken, this procedure will continue to be of considerable help in the feeding of the premature infant without the hazards mentioned above.

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PRE AND POST OPERATIVE PSYCHOLOGIC CARE OF THE PEDIATRIC PATIENT

Special Report No. 255

Charles R. Webb, M.D.

It is the cold, grey dawn. A child is awakened and hurriedly dressed by his tense, anxious parents. In answer to his bewildered questions, he is given a vague or misleading explanation of his destination. Perhaps they tell him he is going on a pleasant jaunt to the zoo, or to see a favorite relative. Sometimes he is answered with the harsh, naked truth: "You're going to the hospital to get your tonsils cut out!"

During the ride to the hospital, he becomes either more and more fearful and distrustful or it becomes increasingly evident that this is not a trip to the zoo or to "Aunt Susie's" and he understandably is both enraged and terrified when he finally arrives. On the other hand if he has been told that he was going to be "cut," the trip merely gives him time to increase his fright as he conjures up fearful fantasies about what will be done to him.

On arrival, he is herded together with other unfortunates, in varying degrees of panic, and is given a quick physical examination by a somewhat disgruntled physician. Other people jab him with long needles and then lead him off to a barren white room where he is further humiliated by being undressed again. Still other white clad strangers forcibly separate him from his mother, put him on a cart and wheel him down the long, sinister appearing corridor. A door opens, he smells strange odors, and, as he is wheeled into the operating room, he catches a glimpse of many sharp, shiny, destructive instruments and of masked, white clad figures.

While his terrified mind is trying to grasp the meaning of all this, his face is covered and he experiences a suffocating sensation as the sickening, sweet "ether" fumes are administered. He tries to struggle, but despite his frantic efforts, feels himself falling, falling into oblivion.

When he awakens, he hurts, he is nauseated and he is bleeding! Once again he is in a strange room, and if he is fortunate, his tearful mother is there to offer some measure of comfort.

"What has been done to me?" he wonders.

"Why has it been done?"

These are the main thoughts that seem to flash through his mind. Thus does a child undergo a "minor operation."

A dramatic description? Yes. Overemphasized? No, for this sequence of events, repeated in varying details, is performed everyday in thousands of hospitals on thousands of children. These pediatric cases may be there for

the so-called "routine" tonsillectomies, circumcisions, or appendectomies; for emergency operations or for diseases of a more serious nature requiring extensive surgery, mutilating techniques or amputations. The important thing is that they are there and are going to be operated upon.

It is the purpose of this paper to examine the feelings and reactions of these patients and the role played by their parents and the medical personnel involved. Further, we will discuss the need for adequate handling of these emotions and their expressions and will offer recommendations for accomplishing this.

From the child's viewpoint, surgery represents an attack upon his person. As has been pointed out by Dr. Adelaide Johnson⁽¹⁾, the child, early in his development, forms a mental image of his entire body and incorporates his impressions of the various parts into his total feeling. He regards the individual members with as much affection and appreciation as he does the whole. This concept, or body image, as it is termed, is fortified or modified by the attitude displayed by his parents towards his body, and its appendages; by incidents and performances that create emotional impressions about it, and by any trauma that may occur to it.

When he is operated on, especially by a mutilating procedure, the patient experiences fear of loss of the part. As has been brought out in analysis, he feels disgust as well as rage at the alteration in his concept of his body image. Also, he is anxious about this change as seen by others, and, he has fears about what has happened to the lost member. This has been forcibly demonstrated by Kolb, Frank, and Watson⁽²⁾ in discussing the treatment of patients with phantom limb fantasies. It takes a long period of analysis to obtain this information and the subsequent emotional response.

This same concern about the lost part and its effect on the personal integrity of the patient has been noted not infrequently in interviews with children who had appendectomies or tonsillectomies.

As a result of the surgical assault, therefore, the child experiences fear and rage. His fear is two-fold, not only the fear of loss of a body part, but also of separation from his parents. His anger is directed toward his parents for permitting the outrage to occur and toward the hospital personnel for committing it.

Further, his anxieties may stir up emotional conflicts and involve his guilt feelings also. As a result, the operation frequently represents punishment for real or imagined misdeeds, mutilation by the parents, and in cases of the seriously disturbed child, it is equated with an attempt at his total annihilation.

Although the child may feel fear and anger how does he react? The overt expressions of his feelings depend on several factors: The age of the

patient, his previous emotional adjustment and security, the emotional equilibrium of his parents, and the type of preparation for surgery are all important determinants in his response.

Frequently he suppresses all his emotions concerning what is about to happen or has happened. His feeling may not be manifested directly, but rather as changes in behavior. In this regard Levy⁽³⁾ pointed out that in the 124 behavior problems he reviewed, 24 of them, representing 20 per cent, had emotional sequelae as a result of an operation. These reactions were manifested by night terrors, fears, especially of the dark, dependent needs, and negativism.

The largest percentage of patients with resultant behavior problem lay in the groups who were under three years of age. Sixteen children of the 25, representing 64 per cent of the group, were in this category. He felt that these patients also had a greater intensity of response to the stimulus of surgery than did the older ones. The higher incidence in this group is attributed to the fact that their "security in life is invested to the highest degree in home and mother."

Levy emphasized that they were less well equipped to handle the anxiety which Dr. Edith Jackson⁽⁴⁾ described as the resultant of any child's exposure to the strange and frightening environment of the hospital in which he was subjected to painful manipulations of his body.

The younger children are more dependent on their mothers, have less experience in social contacts and in knowledge of the world and have little comprehension of what is to ensue.

Unlike older children, they do not possess the skills and concepts to play out, alone or with others, the fearful event which is about to occur or has transpired. Further, because of their limitations in experience and comprehension, it is felt that it is difficult to explain to them what the operation is all about.

While, admittedly, these children were a selected group, insofar as they were all referred to Dr. Levy for behavior problems, the data is felt to warrant the conclusion that these reactions would not differ widely from a normal sampling. Indeed, prior to surgery, 13 of the 25 had no evidence of difficulty.

Dr. Langford⁽⁵⁾ noted that 6 of 20 cases of children with anxiety states were related to the shock of operation. Doctors Lucie Jessner and Samuel Kaplen⁽⁶⁾ took a somewhat different approach to the problem in that they followed a series of 60 children who were brought to the Massachusetts Eye and Ear Infirmary for tonsillectomy and adenoidectomy rather than evaluating selected case material. They attempted to ascertain the elements that might predetermine the reaction of the child to the stress situation, the ways in which the child expressed his feelings, the intensity of this apprehension, and the means utilized to handle these emotions.

It was found that their patients exhibited anxiety with resultant untoward behavior if they were unable to handle their feelings. Of the 60 patients followed, 19 reacted poorly to the experience and 13 others responded in a very inadequate fashion. During subsequent interviews, only 4 of the 19 had improved, and of the 13 who did very poorly in the hospital, there was no reversal in their behavioral trend.

The examiners found that there were three major foci for the children's sources of anxiety: Separation from the home, the anesthesia, and the operation itself. Under the age of 4, the separation from the parents with resultant expressions of loneliness and lack of protection were manifest. The older children showed more fear of the anesthesia and of the surgical procedure which they equaled with mutilation and punishment. The fear of losing control under the anesthetic and the attendant sensation of an overwhelming helplessness was most evident in those patients between the ages of 6 to 12 years.

The anxieties revolving about these foci were expressed by means of four general patterns:

- (1) Obvious terror and panic.
- (2) Depression.
- (3) Symptom formation, which was displayed by nightmares, restlessness, hyperactivity, immobility, anorexia, diarrhea, frequency of urination or various behavior problems.
- (4) Attempts at masking the anxiety. This was done by utilizing such defenses as denial that an operation had been performed; projection of the procedure onto a doll or another person; overcompliance to hospital routine, repression, displacement, and identification with other children or with the aggressor who is, of course, the surgeon.

An interesting defense mechanism we will discuss later in relation to the parents and the doctors concern the technique of overcompliance and of "being good."

In evaluating their patients they classified them as responding in an adequate or poor manner. Adequacy was based on the patient's indication of his awareness of the reality situation and his ability to express his feeling through play or by verbalization. The poor reactors were those who were felt to be unable to utilize either fantasy or reality in order to face the actuality of the situation.

Of those children who were considered to have an adequate response, 22 of the 28 were evaluated as having a healthy personality, 6 were mildly disturbed.

The moderately poor reactors to surgery included only 4 with a healthy personality, 12 with mild disturbances and 3 who were very disturbed. In the very poor reaction group, none were classified as healthy personalities, 8 were mildly disturbed, and 5 were very disturbed.

This same trend was seen in respect to mother-child inter-relationships. Twenty-seven out of 28 of the adequate responders had a satisfactory relationship. Only 6 out of 19 of the moderately poor response group enjoyed such satisfactory relations. In the very poor response group, only three out of 13 had what was considered to be a satisfactory relationship.

Preparation for surgery demonstrated a similar pattern in that 17 of the 28 adequate patients were well prepared while only 6 out of the 13 poor reactors were well prepared.

Examination of the data revealed the correlation of all these factors observed, in that a good personality structure, healthy relationship with the mother and adequate preparation occurred to the highest degree in those that reacted well. By comparison of the 26 situations in which there were poor mother-child relationships, only 10 were adequately prepared for surgery, and only 1 was considered to have had a healthy personality!

As can be seen from Jessner and Kaplan's study⁽⁶⁾, the basic family relationship is important. If the child already has to cope with numerous unresolved conflicts, the anxiety associated with surgery overwhelms him. Parents, too, have emotions and have anxieties and resultant responses to the event occurring to their child. Even the loving, well-meaning parent can increase concentration of the child's anxieties by exhibiting their own fears at this time. This stimulates the fantasies of the child.

Also, in a misdirected effort to protect him, the parent may try to conceal the coming unpleasant experience. As Vander Veer⁽⁷⁾ points out, this is done in the mistaken belief that they thereby spare both themselves and the child. Actually, the opposite is the case since the child finds himself unexpectedly faced with a dangerous situation for which he has had no psychological preparation.

His vulnerability is doubled by his loss of confidence in his protectors. He reasons: "If they have lied to me once, how can I trust them again?" Naturally this arouses his resentment.

If the parents have been overly strict or given to excessive criticism of the child, he frequently responds by regarding the operation as punishment for misdeeds. Numerous investigations have pointed out the surprising frequency with which young patients state they are sick because they have been bad.

Even if none of these errors have been followed, the preparation may not be adequate to allow the child to expect the sight, smell, sounds, and the general outline of the experience he is to face. In order that parents may achieve effective preparedness, it is frequently necessary that they themselves be advised. This entails the assistance of their doctor.

The family physician should be able to tell the parents that the child must rather be faced by the unpleasant truth rather than a pleasant lie.

The child will accept the operation with less anxiety if he is told in general what is to be done and approximately what he will experience. He might suggest to them that the child be given an opportunity to verbalize his anxiety or play out these fears. Unfortunately, however, the physician while aware of psychologic implications, too often regards tonsillectomies, appendectomies and the like as "minor" or "simple" procedures, and he does not pause to consider what the child feels about them. In the instances of what he considers "serious", extensive or mutilating surgery, his attention and that of the surgical staff are directed primarily to the pathology entailed and the technical details involved.

From the viewpoint of the doctors and nurses, the quiet, passive little patient who bears his procedure, whether it be the loss of a bit of foreskin, or an entire extremity, with the gentle air of a martyr and who tries bravely to "smile through" is the "good" patient. As a practical consideration, from the standpoint of efficiency and handling, this is quite true. But they do not take into account that they represent parent surrogates in the child's mind. Since they represent parental authority, the act of being "good" or overcompliant is, as Kaplan states, a defense mechanism employed by the child to bribe destiny. "Forgetting" the operation is utilized in a like manner. The young patient uses up all his energy and attention in an effort to be perfect according to the parents' standards and their own ideals. The fear and rage they feel are repressed and the child frequently regresses. On the other hand, the so-called "bad" patient may be demonstrating his feelings on a reality basis. He directs his aggressive feelings at the parents or at their substitutes—the hospital staff. The member of the hospital staff who tries to force these children to conform or threatens them, fails to take this concept into consideration.

This paper does not mean to indicate that all children subjected to operation will have residual psychic damage. However, *all* children have feelings concerning what happens to them. Adequate handling of children is necessary to cushion them against the force of these emotions and to help prevent psychic trauma.

Dr. Pearson points out that in a surgical procedure there is the element of psychic shock just as there is physiologic shock. Dr. Coleman, an otolaryngologist, concurs in this belief. From his observations he concludes that psychic trauma is as much a complication as post-operative hemorrhage or infection. Many healthy children survive surgical procedures without the necessity of being administered blood transfusions, oxygen, or intravenous fluids. Their response to them is very gratifying and they are not infrequently employed to hasten convalescence.

Debilitated or seriously ill children, on the other hand, need these procedures in order to ensure recovery. Even so, they may experience a long

convalescence. Some seriously ill children do not recover despite the employment of all the aids in the physician's armamentarium.

So too, the healthy personality frequently survives psychic shock without the help of psychologic support, but this aid however, enables a faster response to the situation. On the other hand, children with varying degrees of emotional problems will benefit from the psychotherapeutically oriented program with fewer traumatic sequelae than ordinarily expected. As in the case of the physically moribund, the severe emotional problems will not be saved by this particular process alone. Also through this method, doctors and nurses will be able to use a more sympathetic attitude towards the children and work without censure or irritation.

An adult is expected to have a mature outlook on life and to react reasonably towards an operation. They have experience in life situations, presumably are able to control their emotions, to channel them into acceptable courses, and to face reality without exaggerated emotional responses. However, we all can recall our own worries about any painful manipulation of our body, despite the apparent insignificance of the treatment. And, adults, not infrequently do experience anxiety and fears out of proportion to the situation they face.

Dr. Erich Lindemann describes the psychologic implications of surgery in adults in his studies which revealed that 13 out of 40 women with no previous emotional difficulties, responded with a "characteristic type of untoward psychiatric reaction resembling a state of agitated depression."

Analysts stress the frequency with which adults in treatment bring up the affective response engendered by surgery performed on them as children, which have been to varying degrees repressed since that time. As Dr. Karl Menninger and others point out, these feelings have influenced the whole personality structure since then.

On this topic, Dr. Helene Deutsch states: "The percentage of analytic patients who have had operations before they came to analysis is extraordinarily high."

She goes on to say, "As a rule the psychoanalytic reliving of an operation which occurred, perhaps years before, is intense and vivid and the reconstruction of the psychic reactions at the time of surgery is especially reliable."

There is agreement among a large number of analysts that an adult's attitude towards an expected operation is determined in a large measure by his childhood experiences and in this regard any operation at early age will play an important role.

It has only been in the last decade or so that some physicians and surgeons other than psychiatrists have taken these same factors into account concerning these young patients.

Dentists who specialize in the care of children have long realized the importance of proper psychologic approaches to their patients. They are not concerned primarily, with sequelae, but rather with the more facile handling of their patient. Cooperation is necessary, since usually a child's teeth are not filled, removed, or corrected with orthodontic devices under the effects of an anesthesia. They cannot rely on the propped-open mouth of an unconscious patient; so in order to achieve the necessary cooperation, they must prepare the child for what is to occur. They allow him to express his feelings and solicit his help. Due to their necessity, they have taken long strides ahead of most of the medical field in this regard.

Anesthesiologists, too, realize the value of adequate preparation of their patients. They note that they obtain better cooperation and an easier induction with fewer physiologic reactions if the child knows approximately what to expect.

An anesthesiologist of my acquaintance has a well deserved reputation in this regard. Technically, he represents the typical well-trained anesthesiologist, further he has realized the implications of psychologic reactions on the part of his young patients.

He sees them before the operation, describes briefly their journey to the Operating Room and then demonstrates his mask to them. He states that the comparison of the ether mask to the oxygen mask of the fighter pilot is well accepted by the air-minded younger generation.

In all applicable major cases, he induces the patients in their own rooms with rectal anesthesia prior to transfer to the operation room. Care is also taken that the children awaken in their own rooms after surgery.

Admittedly, this takes time and patience, but he and his colleagues feel it is worth it in the results they achieve.

Since we are aware of the possibilities, we would be well advised to employ the technique at our disposal to minimize the trauma and prevent, in so far as possible, the sequelae. The following suggestions are made to accomplish the desired results.

In regard to the parents, it is necessary that they be informed of their role by a physician. Parenthetically, an especially helpful booklet has been prepared by the Massachusetts Eye and Ear Infirmary to disseminate this information. The parents should explain to their child, in general what will be done, and should encourage him to verbalize or act out the event and his feelings concerning it.

They can employ a play acting technique to enact the main steps that will transpire. The physical examination, the admittance to a hospital room, undressing, the trip to the operating room, the induction and the return to the room, and the resultant temporary pain are made less frightening in this way.

The child should not be told a lie about his destination. The painful truth is more tolerable in the long run. Also, a frank statement that he may have some pain for a day or so, but that it will pass will be more readily accepted than if the question is ignored. Another word of caution about the choice of terms. It is far better to employ the word "fix" rather than "cut", since the latter conjures up fantasies of loss.

If at all possible, the parent should accompany the child to the hospital, undress him, and be on hand when he awakes. If this cannot be done, the child should be told forthrightly when he can expect the parent. Let me again stress that a lie should never be told and a promise once made must be kept.

The child himself should be treated in a sympathetic and understanding way. When he reacts with rage or fear, healthy expression should be permitted, but limits set so that he will not become alarmed at over-permission.

Further, a child will feel more at ease with some memento of his home. It seems to give him a tangible link to it. A favorite toy, his own robe and slippers are not prejudicial to the necessary order and asepsis of hospital requirements.

Doctors and nurses who appreciate the feelings of their young charges will be able to recognize these mechanisms at work. They will be able to demonstrate tolerance and understanding and will accept the hostility and fear that is reflected on them as parent surrogates.

Sometimes, it is necessary for them to act as the parents in preparing the child for his operation, or to act as a support to their patients in the hospital. These too should be undertaken as a necessary part of their professional role.

The timing of an operation should be given careful consideration. Elective procedures should preferably be postponed until after the age of six. If this is impossible due to pressures from the family, at least this should not be carried out while there are obvious psychic conflicts occurring in the child's environment.

Necessary procedures should be utilized only after the child has had adequate preparation. Even in the case of emergencies, the same preparation is usually possible.

The time of the hospital stay for so-called minor procedures has been subject to scrutiny. It is felt that a 48-hour stay would be advantageous in that the child will be able to acclimate himself to the new surrounding if given a longer period there prior to his operation. The exception to this is in the case of very young children who would have fewer separation anxieties if they went home the same day.

When amputations or other mutilating procedures are to be done, a psychiatrist should be employed to help work through the problems that

arise. The seriously emotionally disturbed child or parent will also need the aid of a psychiatrist to prepare them, follow through hospitalization and watch them afterwards.

As can be deduced from the above recommendations, most of them do not require the services of a psychiatrist. Any physician aware of the emotional elements at work and who will take the time to handle them can render an invaluable service in this regard.

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MENORRHAGIA AT PUBERTY DUE TO IDIOPATHIC THROMBOCYTOPENIC PURPURA

REPORT OF A CASE WITH COMPLETE RECOVERY FOLLOWING SPLENECTOMY

Case Report No. 256

Adrian Recinos, Jr., M.D.
John P. McGovern, M.D.

INTRODUCTION

Excessive menstrual bleeding occurs frequently during puberty. The majority of cases, particularly the moderate ones, are attributed to an ovarian endocrine dysfunction⁽⁸⁾ which is corrected with growth and maturity. Other, more concrete but less frequent causes of menorrhagia in the early adolescent include the corresponding causes of prolonged or profuse bleeding in the adult: damaged endothelium, tumors, polyps, cancer, and blood dyscrasias.

The following case report stresses the importance of considering the organic causes of menorrhagia in any case of unexplained protracted menstrual bleeding, and illustrates the fact that such bleeding may be the first or sole symptom of a thrombopenic purpura.

Case Report

M. K. 47-8436

M. K., a thirteen year old white girl was admitted to The Children's Hospital on the service of Dr. Frederic G. Burke with the following history:

She was apparently in good health until six weeks before admission when she had the onset of her regular menstrual period. This began as usual but the flow was greater and continued for ten days during which time she became quite pale and tired easily. On the tenth day the child was seen by a physician who advised immediate hospitalization. She was admitted to another hospital and was found to have a severe thrombopenia. Treatment consisted of ten or more transfusions, vitamin K, and penicillin, yet the menorrhagia persisted.

Past history disclosed a rather uneventful childhood. The patient had had the usual contagious diseases. She had first menstruated eleven months prior to this illness. The periods, which occurred at four to five week intervals, lasted six to seven days with moderate flow.

Examination on entry to Children's Hospital revealed a well-nourished and well-developed girl whose appearance was of the stated age. The skin and mucous membranes were pale and there were a number of petechiae on the gums and over the left abdomen. There were many needle puncture marks and ecchymotic areas on the arms and hands. Bright red blood was oozing from the vagina. The heart, lungs, and abdomen were apparently normal and the liver and spleen were not palpable.

A blood count taken on admission disclosed 10.5 grams of hemoglobin; 3,600,000 red blood cells; 3,700 white blood cells with 57 per cent neutrophils and less than 10,000 platelets per cubic millimeter. Examination of the aspirated sternal marrow

on the second hospital day revealed a hyperplasia of the nucleated red cells, a normal number of megakaryocytes, but a marked reduction in thrombocytes. The white cells appeared in normal proportion. There was no clot retraction after thirty-six hours. The patient's blood group was IV-O, Rh negative.

The child was given several blood transfusions without alteration of the vaginal flow. On the fifth hospital day, a splenectomy was performed. At the beginning of the operation there was profuse bleeding from the skin and subcutaneous tissues. This subsided shortly after the pedicle was clamped, however, and ceased altogether in four days. The platelet count twenty-four hours after splenectomy was 50,000 and in forty-eight hours reached 100,000. Four days postoperative it was above 300,000 and remained normal. The patient improved in color, strength, and spirits and was discharged apparently in good condition on the fourteenth hospital day.

The child remained well and resumed a normal menstrual cycle. Follow-up observations are limited because the family moved to the far-west. However, the child was in good health and had had no abnormal bleeding when last contacted two years after splenectomy.

DISCUSSION

Hamblen⁽²⁾ emphasizes that irregularities of uterine bleeding may be vagaries of normal menstrual physiology, symptoms of diverse diseases, or disturbances of function.

The etiology of menorrhagia, the dysfunction represented by our case, may be classified in three main compartments: general or constitutional conditions, local pelvic lesions, and endocrinal dysfunction. The following outline is modified from Hughes⁽³⁾.

ETIOLOGY OF MENORRHAGIA

A. General or Constitutional Conditions

1. Malnutrition
2. Blood dyscrasias
3. Cardiac decompensation
4. Hypertension
5. Psychological causes

B. Local Pelvic Lesions

1. Uterine fibromyomata
2. Polypi
3. Uterine displacements
4. Retained products of conception
5. Subinvolution after childbirth
6. Carcinoma of the cervix
7. Inflammatory lesions of the Fallopian tube and ovary
8. New growth of ovary
9. Syphilis of ovary

C. Endocrinal Dysfunction

There are numerous reports⁽⁴⁾ in the literature associating menorrhagia with the blood dyscrasias. We were able to find cases in which the menorrhagia was the *presenting* and only symptom of a thrombopenic (idiopathic

thrombocytopenic purpura, aplastic anemia, or leukemia^(5, 6, 7, 8). Three of these cases occurred in the pubertal period, and, like our own, were associated with an idiopathic thrombocytopenia. Complete recovery was reported following splenectomy in one severe case⁽⁸⁾ which closely paralleled our own. Spontaneous recovery undoubtedly has occurred many times in more moderate cases of idiopathic thrombocytopenic purpura. In fact such a case was reported in 1775 by Werlhof (cited by Dameshek et al.^(6, 9) with the first definite description of the disease.

In a study of Barnes⁽⁴⁾, it is of interest to note that of 60 menstruating women with blood dyscrasias, 12 had menorrhagia. Each of these, however, presented sufficient evidence to justify the increased bleeding on a strictly gynecological basis, functional or organic, irrespective of the blood picture. He, therefore, suggests the possibility that other mechanisms exist whereby the patient with hematological bleeding tendencies acquires hypermenorrhea.

Although the total number of cases of excess uterine bleeding associated with blood dyscrasias is relatively small, the findings in the literature and in our case certainly warrants early complete blood studies in every unexplained case of menorrhagia.

SUMMARY

1. A case of menorrhagia at puberty caused by idiopathic thrombocytopenic purpura and apparently cured by splenectomy is reported.
2. The etiologic factors in menorrhagia are outlined and a brief review of other cases of excessive menstrual bleeding associated with thrombocytopenia is presented.
3. The importance of complete blood studies in any unexplained case of menorrhagia is stressed.

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CLINICO-PATHOLOGICAL CONFERENCE

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June R. Pollack, M.D.

A 13-month old white female, L. B. 50-8469, was admitted to The Children's Hospital on July 16, 1950, with complaints of diarrhea of three months' duration. Her early developmental history was entirely normal until she was 10 months of age, at which time she began to have watery, foul-smelling stools. At first intermittent, they soon became constant, and were described by the mother as containing undigested food particles and black sand-like material, and occurring two or three times daily. There had been no associated vomiting or melena. She weighed twenty-two pounds just before the onset. Progressive weakness, semi-stupor, and inability to hold her head up prompted the mother to seek hospitalization.

Further questioning revealed that the mother had sought medical aid two months before, when the diarrhea became constant, and she noticed a protuberant abdomen and thinning of the arms and legs developing. Elimination diets had had no effect on the character of the loose stools, and stool examinations for parasites had been fruitless. Concurrently, pus had been noticed in the eyes each morning, and her upper lids drooped almost constantly except just after awakening from a nap. The child desired an increasing number of naps prior to admission, and her activity diminished markedly. There had been marked weight loss; she had had two bouts of mild upper respiratory infection in the six months before hospitalization, no other illnesses were elicited.

The details of her feeding history were as follows: she was breast fed for the first three days of life, and then switched to an evaporated milk, water, and Karo syrup formula. She spit up frequently while taking this, and at age two months, was given boiled homogenized milk, water, and Karo syrup, which she tolerated well. Boiled whole milk was begun at six months, and after age nine months until admission she received whole milk with the exceptions noted below. She received five drops of Drisdol and the juice of one orange daily from three weeks of age, although 50 milligrams of ascorbic acid was substituted for the orange juice two months before admission. Pabena was the first solid food given, starting at five weeks of age. Pureed fruits at two months, pureed vegetables at three months, egg yolk at four months, and strained meats at five months followed, and all were tolerated well.

When the diarrhea began at age ten months, all foods were withheld for three days and only gingerale, tea, and water were given. Powdered protein milk, boiled skimmed milk, and various combinations of foods were tried in the next week without abatement of the diarrhea, and after several attempts at special non-allergic diets, the mother gradually returned to feeding her a normal diet. A rash was noted about the mouth at one time, which was attributed to soft-boiled eggs, but never recurred when hard-boiled eggs were substituted. No allergy tests were run, although allergy was strongly suspected; the father had had hay fever, and was known to be sensitive to trees and grasses.

This thin, pale, chronically ill, emaciated, little girl was lying listlessly in bed at

physical examination. Her weight was sixteen pounds, fourteen ounces. The skin showed marked loss of subcutaneous tissue, and it hung in many folds. Her hair was thin, sparse, and straight; a small, soft mass near the anterior fontanelle was thought to be a sebaceous cyst. Both upper lids of the eyes appeared swollen and drooping, with conjunctival injection and a slight purulent discharge. The tongue was moist and white-coated, but there was no glossitis or geographia. The ears, nose, and throat, and chest showed no signs of infection. There was a soft, blowing, systolic murmur heard in the third left intercostal space parasternally which transmitted to the neck, and there was no thrill associated. The abdomen had the typical pot-belly appearance of a celiac, and superficial veins were prominent. Cervical, axillary, and inguinal nodes were easily palpable, probably due to the loss of tissue. The extremities were thin and showed poor muscle tone, but were not atrophied. Neurologic examination was negative.

The patient's initial work-up was directed toward establishing some disturbance or disease of the gastrointestinal tract as the cause of the diarrhea, with celiac disease as the working impression. At the same time possible metabolic disturbances were investigated. As test after test was performed, and no light was shed on the problem, various clinical trials on therapy were tried. The foregoing are summarized below:

1. Gastrointestinal Work-up.

A. Infectious:

Stool cultures for pathogens (7/17, 7/18, 7/19, 8/24) were negative.

Stool for ova and parasites (7/25, 8/30) were negative.

Complement fixation test for amebiasis (8/24) was negative.

Complement fixation test for leptospira (8/24) was negative.

Histoplasmosis skin test was negative (8/11).

B. Celiac Syndrome:

Trypsin in stools (7/25, 7/26, 7/28) was negative, and (7/28) was positive in a 1:5 dilution.

Duodenal drainage (8/3) showed a normal content of trypsin.

Stools for fats and starches (8/16, 8/30) were normal.

C. Allergic:

Skin tests to 12 common foods were all negative.

On numerous differential counts of white blood cells—three eosinophils on one occasion only (9/4).

Clinical trials—See below.

D. Roentgen:

Gastrointestinal series (8/7) was normal.

Barium enema (8/6) was normal.

Proctoscopy (8/29, 9/7) was normal; scope advance, six inches.

2. Metabolic Work-up.

Carbon dioxide combining power (7/18) showed 39 volumes per cent and (9/16) 54 volumes per cent.

Blood urea nitrogen (7/17) 15 milligrams per cent.

Non-protein nitrogen (8/16) 39.5 milligrams per cent.

Plasma chlorides (7/17) 585 milligrams per cent.

Calcium (7/17) 12.7 milligrams per cent; (9/1) 12.8; (9/8) 8.5; (9/16) 10.2.

Phosphorus (7/17) 3.2 milligrams per cent; (9/8) 4.7 milligrams per cent; (9/16) 5.1 milligrams per cent.

Total proteins (7/17) 6.50 grams per cent.

Albumin 5.20 grams per cent, globulin 1.30 grams per cent, and A/G ratio 4.1:1.
Thymol turbidity (8/11) 2.8 units.

Cephalin flocculation (8/11) negative at 24 hours and 2+ at 48 hours.

Cholesterol (8/16) 160 milligrams per cent.

Sodium (8/15) 313 milligrams; (9/16) 294 milligrams per cent.

Serum lipase (8/16) 0.05 cubic centimeters of N/20 NaOH/1 cubic centimeter (normal).

Glucose Tolerance Test:	(8/14)	(9/1)
Fasting	100 mgm. per cent	110 mgm. per cent
½ Hr.	195	215
1 Hr.	340	220
2 Hrs.	125	120
3 Hrs.	105	80

Alkaline phosphatase (9/8) 4 Bodansky units.

Eosinophil counts (8/15) 321 cubic centimeters; (8/22) 162; 150; 102; and (8/23) 72; 51.

3. Clinical Trials.

Elimination diets—starvation, goat's milk, nutramigen, denatured diets, celiac diet, pancreatic granules (tried for various periods of time during entire hospital stay)—no effect on diarrhea.

Antibiotic—chloromycetin, kapectate and thyroid extract (8-12 to 8-19) no effect on diarrhea.

Addition of concentrated vitamins, iron, liver extract, and B₁₂. No effect on diarrhea.

Prostigmine—(7-17 to 7-19). No effect on lid drooping or lassitude.

4. Miscellaneous Work-up.

Complete blood counts and urinalyses were always within normal limits.

PPD #1 and 2—Negative.

V.P.C. (7/17) 39 centimeters.

Bone marrow (8/19) was normal.

Stools for occult blood (7/22, 8/29) were negative.

Spinal fluid examination (8/18) normal.

Chest x-ray (7/16) normal, as was the skeletal survey (7/16).

Intravenous pyelogram (8/12) showed a normal urinary tract.

At this examination, however, the x-ray films of the gastrointestinal tract series (8/7) were reviewed and the following reading was added: "A review of this and all other films shows an area of speckled calcification about one inch in diameter in the right vertebral gutter between the tenth and eleventh thoracic vertebrae. The nature of this lesion is not known. A calcified cyst, adrenal tumor, and teratoma should be ruled out."

Flat plates of the abdomen (8/17, 8/29) gave confirmation of speckled calcification.

From the galaxy of laboratory data presented above, it is obvious that this case baffled those connected with it. Thorough investigations of the possibilities of infection, coeliac disease, mucoviscidosis, and allergy as the cause of the diarrhea showed no laboratory evidence to support any of these as the diagnosis. As the diarrhea continued unabated during the hospital course (even during a three-day period when three drams of kapectate were given every four hours), clinical trials at treatment for these diseases were tried; failure to respond only bore out the laboratory findings. The numerous blood chemistries were all within normal limits. Proctoscopic examination performed on two separate occasions revealed a normal mucosa.

The first positive finding was noted on August 12, 1950, when the speckled calcification in the adrenal region was seen on x-ray examination. Investigation of this mass prompted the intravenous pyelography, eosinophil tests, glucose tolerance tests, numerous calcium, and other blood chemistry determinations, and blood pressure determinations, all of which were within normal limits. Although the adrenal gland was strongly suspected of being the site of pathology, it could not be directly or indirectly incriminated; one clinical feature which became evident was frequent and sustained flushing of the face.

On August 19, 1950, an eye consultation revealed that there was a duplication of the normal single row of lashes (dystichiasis) with an associated blepharoconjunctivitis and some type of dermatitis of the lids. Saline irrigations cleared the inflammation gradually, and the droopy appearance of the eyes disappeared.

The patient was sustained medically for the first 58 days of hospitalization and in spite of all measures tried, she went downhill gradually, weighing fourteen pounds, seven ounces on September 11, 1950. The diarrhea continued. Physicians at the hospital were faced with an undiagnosed patient in whom only one abnormality had presented itself, and no one could associate the symptoms of diarrhea with a mass in the right adrenal region. When all else had failed, it was decided to operate on the patient and accordingly, on September 12, 1950, an exploratory laparotomy was performed.

General survey of the abdominal contents showed no abnormality. In the region of the right adrenal gland there was a nodular, firm, rubbery tumor, measuring approximately 4 x 3 x 2 centimeters, which appeared to have a thin layer of adrenal cortex encapsulating it. There was no evidence of local infiltration of the tumor and there were no regional lymph nodes. The mass was shelled out, and the child returned from the operating room in good condition. The surgeons believed they had removed a neuroblastoma.

The patient tolerated the procedure well, and on the third post-operative day a normal stool was reported. From that time on, the stools remained normal, and she was placed on a regular diet. She was discharged on the eleventh post-operative day dramatically improved, having gained two pounds, and the flushing of the face had disappeared. Microscopic sections of the tumor showed it to be a benign ganglioneuroma. Follow-up observations over an eighteen-month period have revealed a perfectly normal, healthy girl of average size. There has never been another diarrheal stool noted.

DISCUSSION

John P. McGovern, M.D.

I believe, if one should have encountered this patient at the bedside as the case unfolded, that his thinking would have been divided into two distinct compartments. The first would incorporate the course and the results of his findings preceding the roentgenographic demonstration of the area of calcification in the region of the right adrenal gland; the second would represent the modifications in reasoning engendered by this observation.

Possibilities that arise with fragmentary notations of significance are listed in outline form within these two stages. Limitation of space prohibits a detailed analysis and justification of each thought.

STAGE I (Before the finding of calcification in area of right adrenal gland)

A. Specific involvement of the gastro-intestinal tract.

1. Bacterial enteritis—Three negative stool cultures for pathogens.
2. Helminthic infestation—Stools negative for ova and parasites; negative for occult blood (hookworm) and past history help rule this out.
3. Amebiasis—Negative complement fixation test and absence of occult blood, against; no warm stool properly examined, however, and cannot rule out amebiasis definitely at this stage with information given.
4. Gastro-intestinal allergy—Several courses of elimination diets, non-revealing; series of skin tests, negative.

B. Celiac syndrome.

1. Mucoviscidosis (fibrocystic disease of the pancreas)—Against would be: age of onset, consistent watery character of the stools, normal amount of trypsin by duodenal drainage, normal serum amylase, lack of lung involvement by this age, normal stool fat, normal gastro-intestinal series and lack of anemia.
2. Celiac disease—Against would be: consistent watery character of the stools; normal stool fat and starch; normal serum amylase, cholesterol and glucose tolerance tests.
3. Starch intolerance—Against would be: the marked hydrolability, severity, normal stool starch and normal serum amylase.
4. Infantile steatorrhea—Against would be the age of onset and the normal stool fat.

C. Tuberculosis (with or without gastro-intestinal involvement)—Against would be the negative PPD, x-ray of chest, and lack of contact by history. Cannot rule out, however, as could have anergic response to PPD at this age, negative x-ray of chest, and no history of contact and *still* have T.B.C. of intestinal tract.

D. Histoplasmosis—Negative skin test against, but certainly does not rule out (see T.B.C.).

E. Myasthenia gravis—No response to three days of prostigmine; extremely rare in infants.

STAGE II (After the finding of calcification in area of right adrenal gland)

A. Tuberculosis with involvement of one or both adrenal glands (Addison's disease)—Against would be the findings given before under tuberculosis; also relative rarity in children, there having

been 53 cases reported in infancy and childhood through 1946⁽¹⁾. Strongly in favor, however, is the highly suggestive course of asthenia, anorexia, progressive weight loss, dehydration, general debility, and gastro-intestinal symptoms with prolonged diarrhea. Records as to the presence or absence of pigmentation are not found in the protocol. The classical laboratory findings in adult Addison's disease have been shown to be quite variable in the reported cases occurring in infancy and children⁽¹⁾. Their presence or absence is, therefore, of considerably less value in the differential diagnosis. The relatively low CO₂ combining power of 39 volumes per cent on admission, however, might reflect a low sodium. Sodium determinations were not done until after parenteral fluids had been given so that the sodium level of 313 mg. per 100 ml. would be of little help. The chloride of 585 mg. per 100 ml. on admission was at the lowest limit of normal and would fit the pattern as would the 39.5 mg. N.P.N. One would have liked a potassium level on admission, 17 keto-steroids, and adrenal function tests. Although the fasting sugar and glucose tolerance tests were within normal limits, low glucose values were found to be the most variable of the accepted laboratory findings in Addison's disease when it occurred in infancy and childhood.

B. Tumor of the right adrenal gland.

1. Cortical tumor—Against is the absence of virilizing or feminizing signs.
2. Medullary tumor:
 - a. Sympathogonioma
 - b. Sympathoblastoma
 - c. Ganglioneuroma—Rare and gives rise to no symptoms.

Calcification frequent in reported cases, however.

C. Pheochromocytoma—Characteristic symptom—complex absent.

Of the diagnoses considered, certainly none completely or adequately explains the course of this disease, with the signs and symptoms both present and absent. Had I not previously observed and recalled this case and *a priori* known that it was a ganglioneuroma of the right adrenal gland, I would have given the final diagnosis as follows:

1. Chronic adrenal insufficiency of the addisonian type (tuberculosis of the adrenal gland).
2. Ganglioneuroma of the right adrenal medulla.
3. Histoplasmosis.

I might add that in 1870 Guttman⁽²⁾ presented a case of a girl, aged 11

years, who was debilitated for one year and died suddenly with gastrointestinal symptoms predominating. At autopsy, the right adrenal was found intact, and the left adrenal contained a large calcified focus. Several other cases of Addison's disease in childhood are reported in which one or both adrenal glands were involved with a tuberculous process, yet there was no other evidence of tuberculosis.

PATHOLOGIC DISCUSSION

David F. Bell, Jr., M.D.: The student's diagnoses are as follows: neuroblastoma, fibrocystic disease of the pancreas, tuberculosis of the adrenal gland.

If there is no further discussion we will have the findings at laparotomy.

June R. Pollack, M.D.: At laparotomy, a right rectus incision was made and exploration of the right adrenal gland revealed a rubbery, firm, nodular, pink tumor with a thin strip of yellow tissue resembling adrenal cortex at its superior pole. The tumor was adherent to the right kidney but was well encapsulated and was removed together with the strip of adrenal cortex. The left adrenal gland, both kidneys and the liver showed no abnormalities and the nodes draining the area of the right adrenal were not enlarged.

The surgical specimen measured 4 x 3 x 2 centimeters, was pink, rubbery and nodular and on section revealed bulging nodular pink tissue.

Microscopic section revealed the small piece of tissue on the superior pole of the tumor to be normal adrenal cortex. The tumor was composed of two main elements: large, differentiated ganglion cells with large eccentric nuclei, and bundles of nerve fibers. Both were imbedded in a stroma of fibrous tissue and Schwann cells.

The girl did well after operation. It was decided to give her no supplemental hormone therapy. Within one week the stools decreased in number to one per day and they became formed, brown and normal in quality. She gained weight, became alert and was discharged. This occurred two years ago and she has been followed at regular intervals. She has gained weight normally and developed normally. There is no lymphadenopathy and no palpable masses can be felt in the abdomen. The liver and spleen are not palpable.

Student: How can a diagnosis of Addison's disease be confirmed in this child when the entire right adrenal gland was removed and no hormone therapy was given?

John P. McGovern, M.D.: It is indeed a point against this being an Addison's disease but certainly this entity seemed to fit the clinical picture.

David F. Bell, Jr., M.D.: It is possible that there may have been a

sympathetic depression of the function of the left adrenal and that after removal of the focus of irritation (ganglioneuroma) the left adrenal cortical function returned to normal. This case seems to illustrate the poor correlation often observed between the anatomical pathology and the clinical response of a patient.

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